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Case Report

Eosinophilic pleural effusion due to *Spirometra mansoni* spargana: a case report and review of the literature

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SUMMARY

A 36-year-old female presented with an eosinophilic pleural effusion. The eosinophilic pleural effusion was considered to have been caused by a parasitic infection. *Spirometra mansoni* spargana was confirmed by semi-rigid thoracoscopy. About 2 months after treatment with praziquantel for 3 days, the pleural effusion had disappeared on the chest roentgenogram.

1. Introduction

Human *Spirometra mansoni* spargana, caused by the plerocercoid of the tapeworm *Spirometra erinaceieuropaei*, occurs mainly by ingesting the raw or inadequately cooked flesh of infected second intermediate or paratenic hosts, such as frogs, snakes, and chicken.[1] The plerocercoids migrate mainly to soft subcutaneous tissues or to muscles, without growing into the adult tapeworm. On rare occasions, they migrate to aberrant sites such as the brain, spinal cord, thoracic cavity, or urogenital organs.[2] Sparganosis patients often show eosinophilia and elevated IgE levels. A case of eosinophilic pleural effusion due to *Spirometra mansoni* spargana, which was confirmed by both histological and immunoserological examinations, is presented herein.

2. Case report

A 36-year-old female was admitted to the First Affiliated Hospital of Wenzhou Medical University in August 2014 with shortness of breath and a mild fever. A physical examination of the patient was normal, except for clinical signs of right pleural effusion. She had eaten cooked frogs 2 years ago, but had never eaten raw meat, swamp crabs, or snakes. A computed tomography (CT) scan of the thorax demonstrated massive right pleural effusion (Figure 1A, B). The initial laboratory data included the following: white blood cell count of 4.59×10^9/l with an increased eosinophil count (14.8%), erythrocyte sedimentation rate of 62 mm/h, C-reactive protein of 32.9 mg/dl, and total serum IgE level of 2123.54 IU/ml. Other laboratory data were within the normal range. The pleural effusion was yellowish in color and contained numerous inflammatory cells, primarily granulocytes (7280×10^6/l), with 54% eosinophils, 12% lymphocytes, 12% neutrophils, and 14% mesothelial cells. Total protein was elevated (66.20 g/l), as was lactate dehydrogenase (2128.00 IU/l) and adenosine deaminase (57 IU/l), but carcinoembryonic antigen (1.5 ng/ml) was within the normal range. Malignant cells were not detected by cytological examination of the pleural effusion. Cultures for bacteria were negative. Thus, a parasitic infection, in particular paragonimiasis, was suspected based on the eosinophilia in the peripheral blood and the pleural effusion. However, neither worms nor eggs were found in the pleural effusion or the stool. Paragonimiasis serology was negative.

Semi-rigid thoracoscopy under local anesthesia demonstrated severe inflammation of the right partial pleura and adhering zone of the fiber bundle, and two off-white bodies were detected in the posterior costophrenic angle (Figure 1C). Following this, a biopsy in this area and another zone of the partial pleura was performed. Histological examination of the wall revealed multiple nodular areas enclosed in fibrous tissue. Gram and acid-fast stains revealed no organisms. The section of bodies revealed a single, small, but

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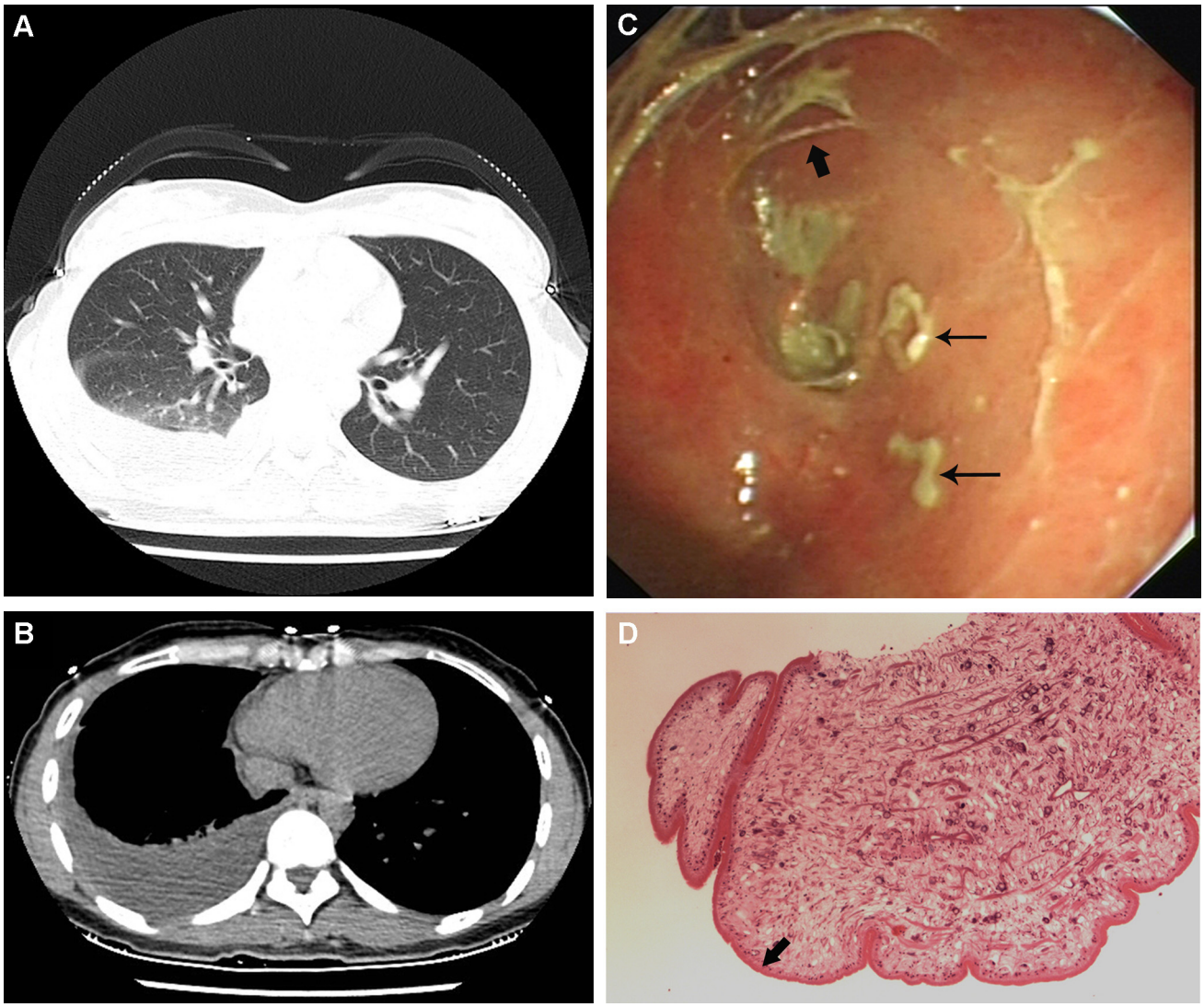

Figure 1. Initial chest CT demonstrated a right pleural effusion (A, B). Semi-rigid thoracoscopy demonstrated two off-white bodies (long arrow), which were detected in the posterior costophrenic angle, and severe inflammation of the right partial pleura and adhering zone of the fiber bundle (short arrow) (C). The sparganum showed a thick eosinophilic uniform tegumental structure (short arrow), and a few calcareous corpuscles and areas of calcification were noted in the body (D).

solid, folded cestode worm. Microscopically, sections of the worm from the case revealed it to have no body cavity and a homogeneously eosinophilic surface tegument that was ridged at irregular intervals; a well-formed scolex was absent. Underlying the tegument, subtegumental columnar cells, longitudinal bundles of longitudinal smooth muscle fibers, and longitudinally arranged excretory ducts in a pale bluish matrix were observed. A few calcareous corpuscles and areas of calcification were noted. The histological features of the worm were characteristic of the larval form of sparganum mansoni (Figure 1D). Meanwhile, the patient's serum was examined by multiple-dot ELISA to detect specific antibody against various parasite antigens, including schistosomiasis, paragonimiasis, strongyloidiasis, cysticercosis, trichinosis, gnathostomiasis, filariasis, Toxoplasma, *Spirometra mansoni* spargana, and toxocariasis. The multiple-dot ELISA was conducted at the National Institute of Parasitic Diseases of the Chinese Center for Disease Control and Prevention (Shanghai). The serum gave marked positive reactions against *Spirometra mansoni* spargana antigen without cross-reactions with other parasite antigens. A chest radiograph showed a right pleural effusion, leading to a diagnosis of pleural sparganum mansoni.

The patient was treated with praziquantel at 75 mg/kg/day for 3 days. The eosinophil count and total IgE level decreased gradually after treatment. About 2 months after treatment, the pleural effusion had disappeared on the chest roentgenogram. The efficacy of the treatment was also evaluated by measuring specific IgG antibody, which was negative after the treatment.

3. Discussion

Sparganum mansoni is the larval form of *Spirometra mansoni*, and causes various clinical symptoms when it parasitizes the human body. The definitive hosts are domestic and wild cats and dogs.[2] Eggs of the worm excreted in the feces of the final host grow to coracidia in fresh water and are eaten by cyclops, the first intermediate host. Each coracidium grows into a procercoid larva in the body of the intermediate host. The cyclops are then eaten by the second intermediate hosts, such as frogs, snakes, and freshwater fish. They pass through the intestinal wall and reside in subdermal tissue and muscle, growing into plerocercoids, i.e., spargana. When the second intermediate host is eaten by the final host, the spargana settle down in the intestinal tract. Humans are generally infected by this worm by drinking freshwater containing cyclops, the first intermediate host, or by eating raw or poorly cooked freshwater fish, frogs, and snakes, the second intermediate

hosts. The present patient had a history of eating cooked raw frogs 2 years before hospitalization, so this was presumed to be the route of infection in this case.

Human sparganosis manifests with various clinical symptoms; it usually presents with slowly growing and migratory subcutaneous nodules, so that patients are mainly diagnosed and treated by dermatologists. The thoracic cavity is a rare site for the localization of this parasite in humans.[3] A literature review identified five reported cases of sparganosis with pleural effusion.[3,4] The median age of these previous cases, including the present case, was 46.8 (range 36–62) years. The case reported here is the first in which the worm bodies were detected by semi-rigid thoracoscopy. Regarding the route of infection to the thoracic cavity, it is proposed that the parasite penetrates the intestinal wall and invades the thoracic cavity through the diaphragm.

Clinical information is not always helpful in making a diagnosis, because human sparganosis shows a great variety of manifestations. In pleural sparganosis, in particular, it is difficult to detect the worms in the pleural cavity, so an immunoserological diagnosis is important for the diagnosis of pleural sparganosis. In the present case, although the worm was found in the pleural cavity, an ELISA test was still used for the diagnosis. Semi-rigid thoracoscopy plays an important role in ascertaining the etiology of eosinophilic pleurisy.[5] In comparison to video-assisted thoracic surgery (VATS), semi-rigid thoracoscopy can been carried out safely under local anesthesia and at a lower cost. Hence, eosinophilic pleurisy patients without contraindications to semi-rigid thoracoscopy should undergo semi-rigid thoracoscopy examination for morphological histopathological evidence as a matter of routine.

Anti-parasitic drugs such as praziquantel and mebendazole may be effective for sparganosis. This was confirmed by the effective treatment of the patient with praziquantel in the present report. However, complete extraction of the larvae is a treatment of choice. As well as being useful for the diagnosis, semi-rigid thoracoscopy is useful for the extraction of larvae in pleural sparganosis.

The majority of patients with sparganosis have been adults, and the symptoms have predominantly been similar to those caused by a space-occupying lesion. Eosinophilia may especially be apparent when the worm is migrating through tissues. Hence the degree of eosinophilia was higher in the pleural effusion than in the peripheral blood.

Although sparganosis is rather rare, this possibility must be considered in cases of eosinophilic pleuritis. Specific antibody detection by multiple-dot ELISA is helpful for the differential diagnosis, and semi-rigid thoracoscopy may be able to locate the worm in the pleural cavity, especially at the costophrenic angle.[5]

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Conflict of interest: None declared.

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